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Case Report

A rare cause of facial nerve palsy in a young infant: Kawasaki disease

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Kawasaki disease (KD) is a vasculitis in which the most common complication is development of coronary aneurysms. Neurological complications rarely occur in KD patients such as facial nerve palsy (FNP). FNP associated with KD may indicate increased risk of coronary artery aneurysm. Infants with facial nerve paralysis and unexplained-prolonged febrile period should be evaluated with echocardiography. Here in, we present a 4-month-old female with FNP and unexplained fever who was diagnosed KD due to echocardiographic findings.

Key words: coronary aneurysm, echocardiography, facial palsy.

Kawasaki disease (KD) is an acute, self-limited panvasculitis of medium size arteries. KD occurs predominantly among infants and young children. Even though it can affect any organ system in the body, the development of coronary artery aneurysms is the most common and life-threatening complication. A lot of different neurological complications can occur in KD patients but facial nerve palsy (FNP) is known as a rare presentation of KD.[1] Furthermore FNP has been reported as a possible marker of more severe disease and have increased risk of coronary artery involvement among infants.[2] In this case report, we present a patient with FNP and unexplained fever who was diagnosed with KD due to a coronary aneurysm on echocardiography (Echo). The family of the case signed an informed consent.

Case Report

A 4-month-old female who presented with 3 days of fever, was admitted to another clinic and hospitalized for a urinary system infection and treated with intravenous antibiotic. Despite antibiotic treatment, her fever continued and facial asymmetry occurred on the 7th day of onset of the fever, and the patient was referred to our neurology department. She had a history of bilateral nonexudative bulbar conjunctival injection which had self-healed within the first few days but she had no cervical lymphadenopathy, polymorphous exanthem, changes in lips and oral cavity, erythema of palms, edema of hands or periungual peeling of fingers. Physical examination revealed fever (39°C), irritability and left sided unilateral peripheral facial nerve palsy. Her laboratory tests revealed anemia (hemoglobin: 7.6 g/dl), leukocytosis (29.500/mm^3), thrombocytosis (718.000/mm^3), increased C-reactive protein level (5.16 mg/dl) and erythrocyte sedimentation rate (105 mm/h), alanine aminotransferase (58 U/L) and decreased serum albumin level (2.5 gr/dl). Cerebrospinal fluid examination and cranial magnetic resonance imaging were normal. The patient was consulted to the pediatric cardiology department because of persistent fever, increased acute phase reactants and unexplained FNP. Echocardiogram demonstrated left ventricle end-diastolic diameter (LVDd): 22 mm (z score: 0.38), LV ejection fraction/fractional shortening: 69%/36%, left coronary artery (LCA): 4.4 mm (z score: 9.51), proximal right coronary artery (RCA): 4.2 mm (z score: 9.51) and coronary artery ectasia at all segments. There was a saccular aneurysm in proximal RCA (Fig. 1). One thrombus which was 4X4 mm in size was seen in the left atrial appendage. Minimal pericardial effusion around the heart and mild

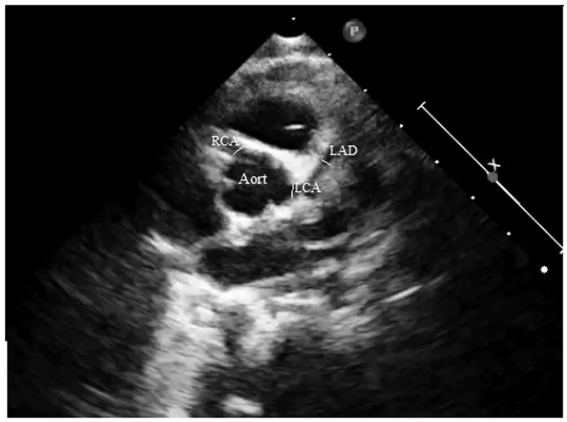

Fig. 1. Echocardiogram before treatment.

to moderate mitral regurgitation with flow velocity 4.1 m/sec was observed. Incomplete KD was diagnosed with her medical history, physical, echocardiographic and laboratory findings. Intravenous immunoglobulin (IVIG) (2 g/kg/12 h) and acetyl salicylic acid (ASA) (80 mg/kg/day), enalapril (0.1 mg/kg/day), and subcutaneous enoxaparin were started. Her irritability and fever resolved on follow-up and FNP recovered 7 days after IVIG treatment. Decreasing of pericardial effusion, mitral regurgitation and resolving thrombus were seen on Echo control (Fig. 2). According to recommendations for the long-term management and surveillance of cardiovascular risk in individuals with Kawasaki disease in the 2004 AHA Guideline[3] our patient was accepted as level III and low dose ASA was continued. In addition, enoxaparin treatment has been continued due to thrombosis. Despite resolution of coronary aneurysms, coronary artery ectasia has continued with minimal improvement at her third month Echo control.

Discussion

Kawasaki disease should be considered in the differential diagnosis of a young child with unexplained fever lasting longer than 5 days and with any of the principal clinical features of this disease.[3]

The term incomplete KD or atypical KD is used for patients who do not fulfill diagnostic criteria (have fever≥5 days but less than four signs of mucocutaneous inflammation). Neurological conditions such as irritability, aseptic meningitis, seizure, ataxia and cranial nerve palsy have been defined at 1%-30%.[4]

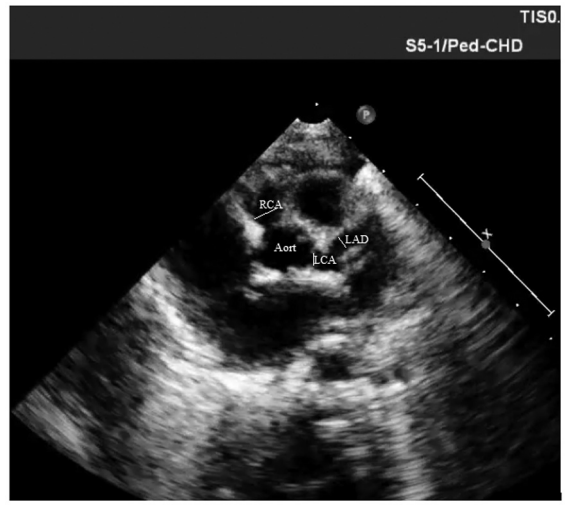

Fig. 2. Echocardiogram after treatment.

But FNP is very rarely seen.[5,6] FNP associated with KD was first reported by Murayama in 1974.[7] FNP is thus thought to be the result of vasculitis involving the facial nerve.[2] Our patient was diagnosed with incomplete KD as they did not fulfill criteria. Although laboratory tests in KD are not specific, they can provide diagnostic support.[3] Our patient has a lot of supportive laboratory findings such as anemia, thrombocytosis, hypoalbuminemia, increased acute phase reactants and intermittent sterile pyuria.

The incidence of central nervous system involvement as ataxia, hemiplegia, seizure and sensorineural auditory loss, in KD ranges from 1.1%-3%. The previous reports claim that KD with FNP may tends to be under 12 months old, more frequently unilateral and on the left, with female/male ratio 1.4/1 (in contrast to the 1.5/1 male predominance for KD, in general), and indicates increased risk of coronary artery aneurysm.[2,6] Because of increased risk of inflammatory burden, KD with FNP patients may require more attentive echocardiography, immediate treatment and additional anti-inflammatory therapy.[6] FNP resolves spontaneously and completely with the duration ranging from 2 days to 3 months, without needing any treatment other than anti-inflammatory treatment of KD.

In conclusion, it is important to suspect for KD in children with unexplained-prolonged febrile period and FNP, to evaluate with Echo carefully. The paralysis normally resolves spontaneously and completely.

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